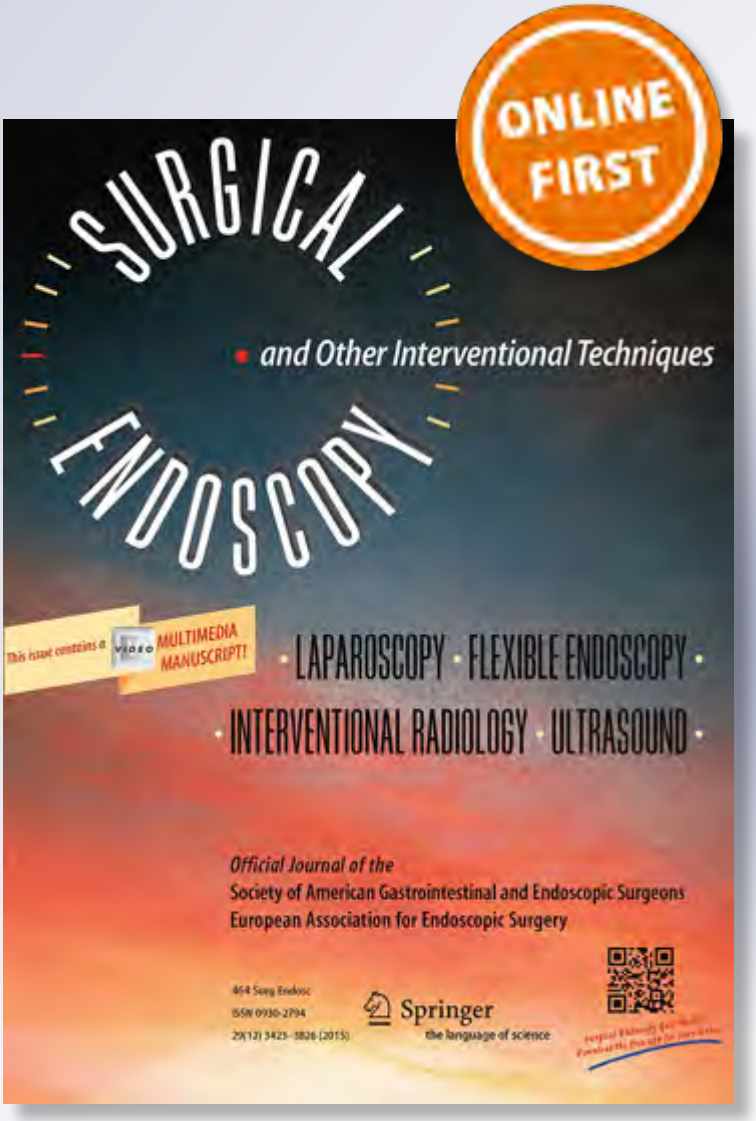
Recurrence of biliary tract obstructions after primary laparoscopic hepaticojejunostomy in children with choledochal cysts

Mei Diao, Long Li & Wei Cheng

Surgical Endoscopy
And Other Interventional Techniques
Official Journal of the Society of American Gastrointestinal and Endoscopic Surgeons (SAGES) and European Association for Endoscopic Surgery (EAES)

ISSN 0930-2794

Surg Endosc
DOI 10.1007/s00464-015-4697-5



Springer

Surg Endosc
DOI 10.1007/s00464-015-4697-5

Recurrence of biliary tract obstructions after primary laparoscopic hepaticojejunostomy in children with choledochal cysts

Mei Diao[1] · Long Li[1] · Wei Cheng[2,3]

Received: 5 September 2015 / Accepted: 17 November 2015

Abstract
Background The aim of the current study was to investigate the cause and develop a management strategy for recurrent biliary obstructions after primary laparoscopic hepaticojejunostomy in children with choledochal cyst (CDC).
Methods Thirty CDC patients (mean age: 7.15 years, range 8 months–24 years, F/M: 22/8) who suffered from recurrent biliary obstructions after primary laparoscopic hepaticojejunostomies were referred to our hospital between January 2006 and June 2014. All patients underwent redo hepaticojejunostomy ± ductoplasty ± reposition of aberrant right hepatic arteries.
Results All patients developed recurrent cholangitis or persistent abnormal liver function 1 month to 7 years postoperatively. Liver biopsy pathology verified that 56.7 % (17/30) of patients had grades I–IV of liver fibrosis. We identified a previously unreported cause of biliary obstruction, i.e., aberrant right hepatic arteries crossing anteriorly to the proximal common hepatic duct in high percentage of the patients who suffered from postoperative recurrent biliary obstructions (7/30, 23.3 %). The hepatic arteries were repositioned behind Roux loop during the redo hepaticojejunostomies. Of remaining patients, nine (30 %) patients had associated hepatic duct strictures and underwent ductoplasties and wide hepaticojejunostomies. Fourteen (46.7 %) patients had anastomotic strictures and underwent redo hepaticojejunostomies. The median follow-up period was 62 months (14–115 months). No recurrent biliary obstruction or cholangitis was observed up to date. Liver functions were normalized.
Conclusions Aberrant hepatic artery, unsolved hepatic duct stricture, as well as poor anastomotic technique, can all contribute to recurrent biliary obstructions after the primary laparoscopic hepaticojejunostomies. Early surgical correction is advocated to minimize liver damage.

Keywords Choledochal cysts · Laparoscopy · Postoperative recurrent biliary obstruction · Hepaticojejunostomy · Children · Aberrant hepatic artery

For children with choledochal cysts (CDC), laparoscopic cyst excision and Roux-en-Y hepaticojejunostomy provide a less traumatic alternative to the conventional open surgery. Similar in principles to the open approach, recurrent biliary tract obstruction is one of the major postoperative complications [1–3]. The current study aims to identify the factors involved and develop a management strategy.

Materials and methods

CDC patients referred to our hospital with recurrent biliary obstruction complication after the primary laparoscopic hepaticojejunostomies between January 2006 and June 2014 were reviewed. All patients underwent redo hepaticojejunostomy ± ductoplasty ± reposition of aberrant right hepatic arteries by the same surgical team. Ethics approval

✉ Long Li
lilong22@hotmail.com

Wei Cheng
wei.cheng@monash.edu

[1] Department of Pediatric Surgery, Capital Institute of Pediatrics, Beijing 100020, People's Republic of China

[2] Department of Paediatrics and Department of Surgery, Southern Medical School, Faculty of Medicine, Nursing and Health Sciences, Monash University, Clayton, VIC 3168, Australia

[3] Department of Surgery, Beijing United Family Hospital, Beijing, China

Published online 10 December 2015

from the Ethics Committee of Capital Institute of Pediatrics was obtained. Written informed consents were obtained from the parents of CDC patients prior to the study.

Preoperative ultrasonagraphic studies, computed tomography (CT) scan, magnetic resonance cholangiopancreatography (MRCP), and intraoperative cholangiograms were carried out to delineate the biliary system. Laparoscopic redo surgeries were carried out as follows: (1) The aberrant right hepatic arteries crossing anteriorly to the proximal common hepatic duct were repositioned behind the proximal hepatic duct during the redo hepaticojejunostomies; (2) patients with unsolved hepatic duct strictures underwent ductoplasties and wide hepaticojejunostomies; and (3) patients with anastomotic stenosis underwent laparoscopic redo hepaticojejunostomies.

Following our intervention protocol, all patients were routinely given antibiotics, glycyrrhizin for the liver function improvement, and ursodeoxycholic acid to facilitate bile drainage postoperatively till the liver function normalized.

Patients were followed up in our clinic 1, 2, 3, 6 months postoperatively and every 6 months thereafter. The liver function tests, ultrasonographic studies, CT scans, and MRCP were carried out.

Statistic analysis

Data were analyzed with SPSS 13.0 package. Paired *t* tests were applied to compare perioperative laboratory values. $P < 0.05$ was considered to be statistically significant.

Results

Thirty CDC patients (mean age: 7.15 years, range 8 months–24 years, F/M: 22/8) who were referred to our hospital for recurrent biliary obstructions after primary laparoscopic hepaticojejunostomies between January 2006 and June 2014 were reviewed.

All patients suffered from recurrent cholangitis with jaundice, abdominal pain, fever, or persistent abnormal liver function 1 month to 7 years postoperatively. All patients underwent conservative treatments with antibiotics, glycyrrhizin, and ursodeoxycholic acid lasting 1 month to 7 years (mean: 2.83 years). Of them, eight patients underwent percutaneous transhepatic biliary drainage for 1 month. Additionally, four patients underwent repeated percutaneous transhepatic biliary drainages for 12–19 months (replaced the drain tube every 3 months) due to the failure of medications.

The mean diameter of stenotic segments was 0.17 cm (0.1–0.3 cm). The mean maximal diameter of dilated proximal hepatic duct was 2.5 cm (2–3.5 cm). Intrahepatic duct stones were detected with ultrasonographic studies, CT scans, MRCP, and intraoperative intrahepatic duct endoscopy in two-thirds of the (20/30) patients. All patients showed significantly elevated serum bilirubin and abnormal liver functions (Table 1). Pathological results showed that 56.7 % (17/30) of patients had grades I–IV liver fibrosis.

Interestingly, seven (23.3 %) patients were found to have aberrant right hepatic arteries crossing anterior to the proximal common hepatic duct, compressing the common hepatic ducts. Preoperative MR showed common hepatic stricture and intrahepatic duct dilatation (Fig. 1A). CT scan before the redo surgery showed compression stenosis in the middle of proximal hepatic duct (Fig. 1B), with no stenosis detected at the site of hepaticojejunal anastomosis. Redo surgeries verified the aberrant right hepatic artery (Fig. 2A). The hepatic arteries were carefully mobilized from proximal common hepatic ducts and repositioned behind common hepatic ducts (Fig. 2B, C). The stones in the intrahepatic bile ducts were completely cleared (Fig. 2B). Redo hepaticojejunostomy was carried out (Fig. 2C).

Nine (30 %) patients were found to have unsolved hepatic duct strictures. Preoperative MRCP, CT scan, ultrasonographic studies, and cholangiograms showed that both extrahepatic and intrahepatic bile ducts were dilatated (Fig. 3A). After the primary surgeries, the imaging investigations demonstrated upstream intrahepatic bile duct dilatation, which was far from the anastomotic sites (Fig. 3B). The anastomotic stoma per se was not stenotic. Redo surgeries confirmed persistent common hepatic duct strictures (Fig. 4A). The ductoplasty and wide hepaticojejunostomy were carried out (Fig. 4B).

The remaining 14 (46.7 %) patients had anastomotic stenosis. Preoperative MRCP, CT scan, ultrasonographic studies, and cholangiograms showed extrahepatic bile duct dilatation without intrahepatic bile duct dilatation (Fig. 5A). After the primary surgeries, the investigations showed biliary dilatations immediately proximal to the anastomotic sites (Fig. 5B). Redo surgeries verified the anastomotic stenosis (Fig. 6A). The stenotic segments were excised. Intrahepatic bile ducts were inspected with endoscope and irrigated with normal saline. The intrahepatic stones were completely removed. Redo hepaticojejunostomies and ductoplasties were carried out (Fig. 6B).

All patients recovered uneventfully. The median follow-up period was 62 months (14–115 months). No mortality or morbidities of bile leak, recurrent biliary obstruction, or cholangitis was observed. Ultrasonographic studies, CT scan, and MRCP showed no intrahepatic duct dilatations after the redo surgeries. Liver function parameters reversed to normal level after surgeries (Table 1).

Table 1 Pre- and post-redo hepaticojejunostomy liver function testes

	Preoperation	Postoperative 2 years	*p*
TBIL (μmol/L) Ref: 3.4–20	158.02 ± 55.78	14.18 ± 4.49	<0.001
ALT (U/L) Ref: <40	328.99 ± 121.10	25.09 ± 9.53	<0.001
AST (U/L) Ref: <40	334.86 ± 140.12	26.22 ± 9.13	<0.001
ALP (U/L) Ref: <400	891.57 ± 332.92	167.97 ± 60.36	<0.001
GGT (U/L) Ref: 7–50	438.47 ± 182.69	29.90 ± 13.32	<0.001

TBIL total bilirubin, *ALT* alanine transaminase, *AST* aspartate aminotransferase, *ALP* alkaline phosphatase, *GGT* γ-glutamyl transpeptidase

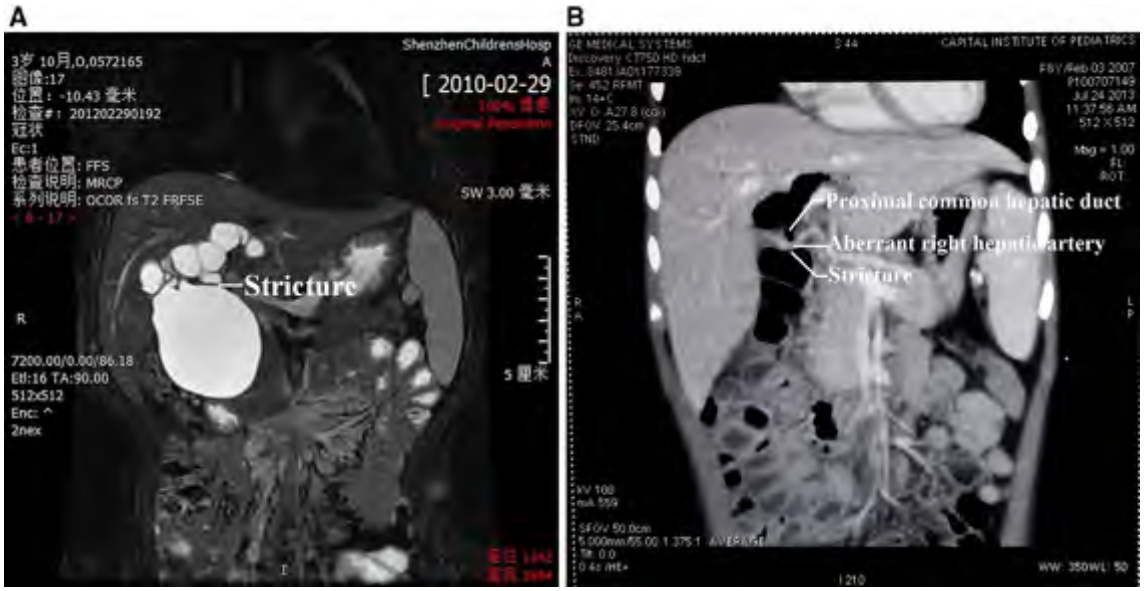


Fig. 1 A 7.5-year-old girl who suffered from recurrent cholangitis since the age of 1.75 years after primary laparoscopic surgery because of compression of an aberrant right hepatic artery compression of common hepatic duct. **A** Preoperative MR showed common hepatic duct stricture and intrahepatic duct dilatation. **B** Postoperative CT showed compression stenosis in the middle of dilated proximal common hepatic duct

Discussion

Our experience demonstrated that aberrant hepatic arteries, unrecognized hepatic duct stricture, and anastomotic technique were the main causes of recurrent biliary tract obstruction after the primary laparoscopic hepaticojejunostomies. We categorized the causes of recurrence into two groups, i.e., congenital and technical:

1. Congenital factor:

    1. Anterior aberrant right hepatic artery: Right hepatic artery anatomy has many variations. The incidence of the anterior crossing right hepatic artery to extrahepatic bile duct ranges from 12 to 24 % [4–6]. Vascular compression at proximal common hepatic duct is a rare congenital cause of obstructive jaundice. Literature has reported that common hepatic duct obstruction can be also compressed by aberrant celiac artery, gastroduodenal artery in addition to the right hepatic artery [5, 7]. Todani [8] firstly advocated to place the right hepatic artery behind common hepatic duct to restore normal anatomy. PichaLal [9] reported that the right hepatic arteries cross anteriorly to the common hepatic duct in 12.8 % (15/117) CDC children. They were repositioned posterior to the biliary-enteric anastomosis to restore normal anatomy. The current study is the first report to identify the aberrant right hepatic artery as one of the major causes of recurrence of biliary tract obstruction after primary laparoscopic hepaticojejunostomy. The mechanism of obstruction is akin to that of aberrant renal artery causing hydronephrosis in the ureteropelvic junction. In choledochal cysts, we speculate that there may be two possibilities: (1) Prior to surgery, intraluminal pressure of dilated biliary system resists the

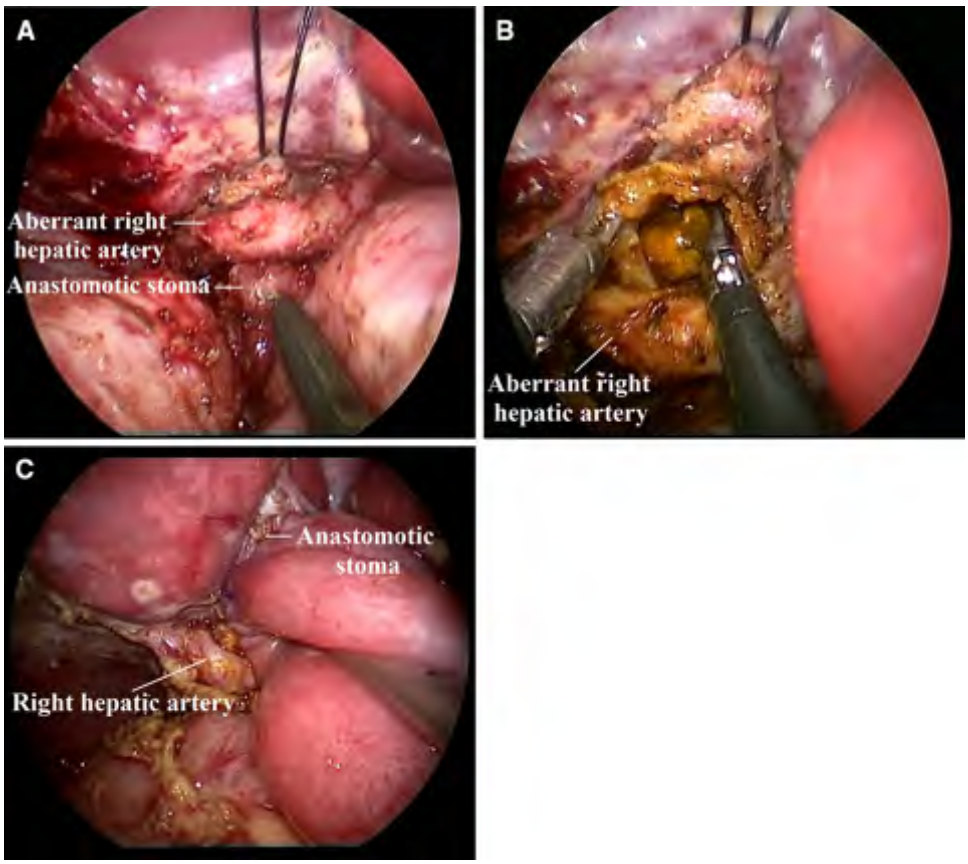


Fig. 2 Laparoscopic ductoplasty and redo hepaticojejunostomy for a 7.5-year-old girl who suffered recurrent cholangitis since 1.75 years after primary laparoscopic surgery because of compression of common hepatic duct by an aberrant right hepatic artery. **A** Aberrant right hepatic artery anteriorly crossing the common hepatic duct. **B** Intrahepatic duct stone clearance. **C** Aberrant right hepatic artery was replaced behind the anastomotic stoma

compression of aberrant right hepatic artery. After surgery, the biliary pressure is relieved. The compression of aberrant right hepatic artery to proximal common hepatic duct is aggravated; (2) the hepaticojejunal anastomotic stoma is close to the aberrant right hepatic artery. In wound-healing process, compression of aberrant right artery to inflammatory edematous common hepatic duct may narrow the lumen. These two factors lead to bile flow obstruction. Our experience demonstrated that repositioning the right hepatic artery behind proximal common hepatic duct does not only to restore normal anatomy, it also prevents postoperative recurrent biliary obstruction and subsequent hepatocellular damage.

2. Unrecognized hepatic duct stricture: Experience and careful analysis of preoperative radiological imaging will prevent it. Multiple strictures of common hepatic and left/right hepatic duct require careful inspections. The stenotic segments should be split to the proximal dilated bile duct. A wide hepaticojejunostomy is then performed at this level.

2. Technical factor—anastomotic stenosis: This usually occurs early in the series of laparoscopic practice, as a result of inexperience. Improvement of anastomotic skill, preservation of blood supply, tension-free anastomosis, and adequate caliber of anastomoses (diameter in older children $\geq$ 1 cm, diameter in neonates $\geq$ 0.5 cm) [10] are the key factors to minimize anastomotic stenosis. In situations in which the proximal common hepatic duct is not wide enough, the anterior wall of common hepatic duct can be incised longitudinally.

Once the postoperative biliary obstruction develops, early surgical correction is advocated. Prior to release of biliary obstruction, the medication which facilitates bile excretion, such as ursodeoxycholic acid, should be used with caution or be avoided altogether. It may aggravate cholestasis, causing further liver damage. Percutaneous transhepatic biliary drainage is not the definitive surgery for biliary obstruction. Repeated percutaneous transhepatic biliary drainage with frequent drain tube replacement may be associated with cholangitis, cholestasis, inflammatory stricture of hepatic duct at the drain site [2, 11]. In our series, two patients with unsolved hepatic strictures underwent repeated percutaneous transhepatic biliary drainages every 3 months. Initially, the dilatation in the right hepatic ducts was more severe than that of the left hepatic ducts. After 1 year of repeated percutaneous

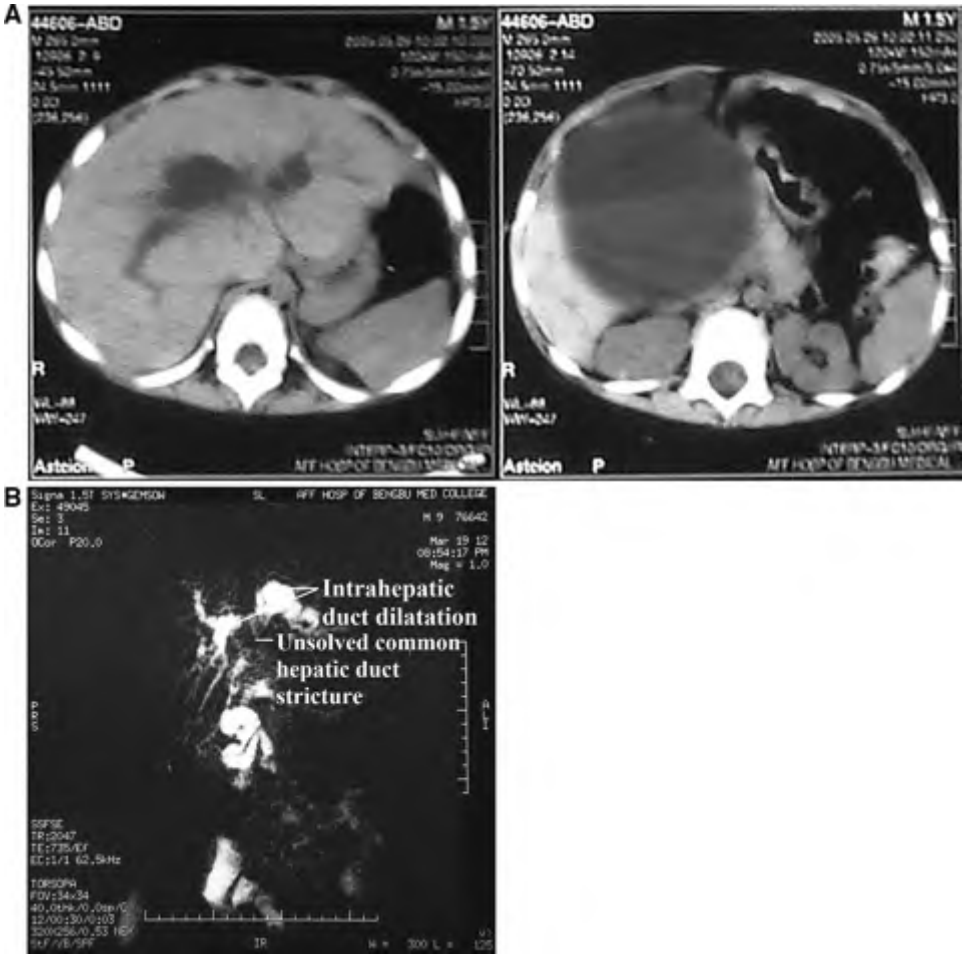


Fig. 3 A 10-year-old boy who suffered recurrent cholangitis for 9 years after the primary definitive surgery because of unrecognized common hepatic duct stricture. **A** Preoperative CT showed both intrahepatic and extrahepatic duct dilatations in a 10-year-old boy who suffered from recurrent cholangitis because of unrecognized common hepatic duct stricture. **B** Postoperative MRCP showed intrahepatic duct dilation

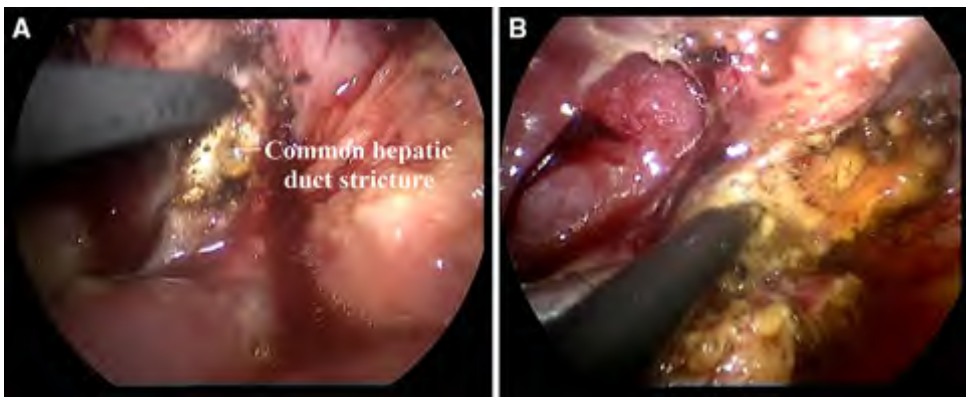


Fig. 4 Laparoscopic ductoplasty and redo hepaticojejunostomy for a 10-year-old boy who suffered from recurrent cholangitis for 9 years after primary definitive surgery because of unsolved common hepatic duct stricture. **A** Unsolved common hepatic duct stricture, **B** ductoplasty for a 10-year-old boy who suffered from recurrent cholangitis for unsolved common hepatic duct stricture

transhepatic biliary drainages through left hepatic ducts, the right hepatic ducts dilatations disappeared, while the left hepatic duct developed inflammatory strictures and severe upstream bile duct dilatation.

In conclusion, aberrant hepatic artery, unsolved hepatic duct stricture, and anastomotic technique are the common causes of recurrent biliary tract obstructions after primary laparoscopic hepaticojejunostomies. All these are preventable by either recognition of the pathologies or technical improvement. Early surgical correction is advocated to avoid severe liver damage.

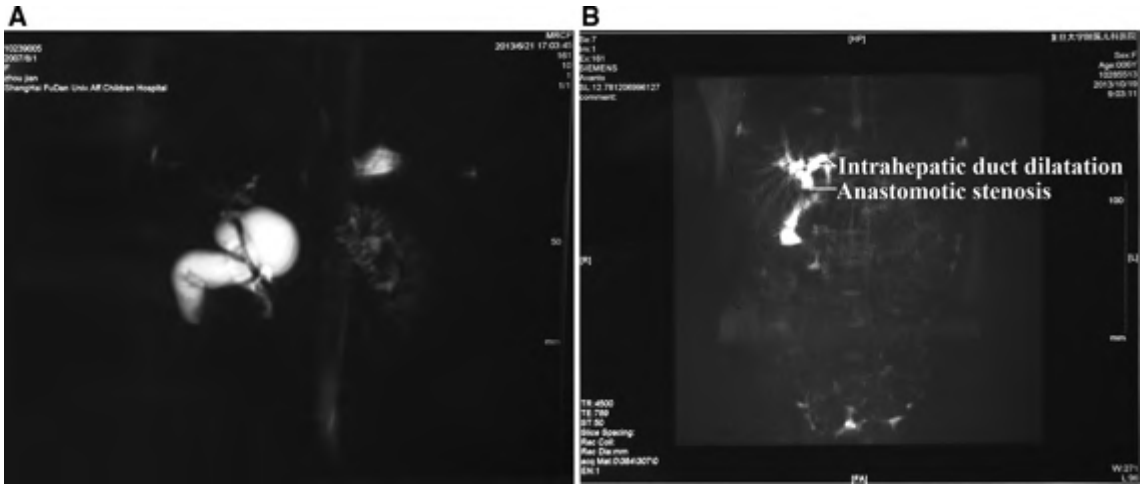


Fig. 5 Anastomotic stenosis in a 7-year-old girl who had persistent abnormal postoperative liver function. **A** Preoperative MRCP showed extrahepatic duct dilatation without intrahepatic duct dilatation in a 7-year-old girl with postoperative anastomotic stricture. **B** Postoperative MRCP showed anastomotic stenosis with intrahepatic duct dilatation

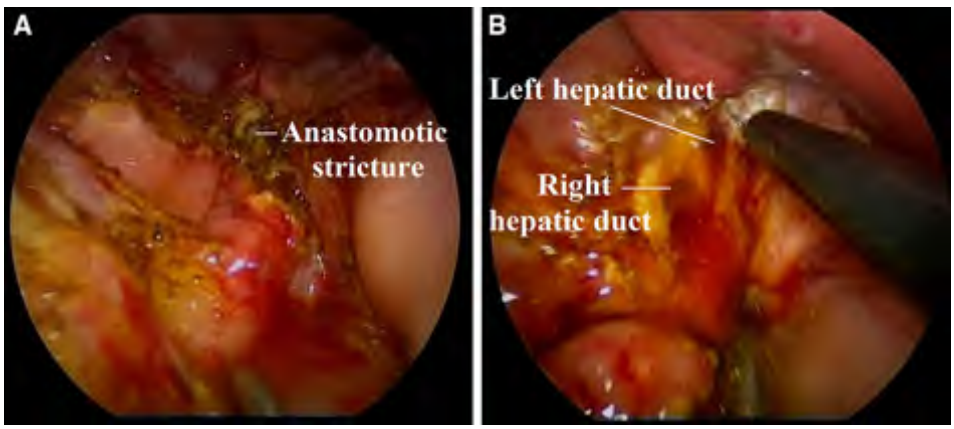


Fig. 6 Laparoscopic ductoplasty and redo hepaticojejunostomy for a 7-year-old girl who suffered anastomotic stenosis after primary laparoscopic surgery. **A** Anastomotic stenosis, **B** ductoplasty of left and right hepatic duct

Compliance with ethical standards

Disclosures Dr. Mei Diao, Prof. Long Li, and Prof. Wei Cheng declare no conflicts of interest or financial ties to disclose.

References

1. Ono S, Fumino S, Shimadera S, Iwai N (2010) Long-term outcomes after hepaticojejunostomy for choledochal cyst: a 10- to 27-year follow-up. J Pediatr Surg 45:376–378
2. Urushihara N, Fukumoto K, Fukuzawa H, Mitsunaga M, Watanabe K, Aoba T, Yamoto M, Miyake H (2012) Long-term outcomes after excision of choledochal cysts in a single institution: operative procedures and late complications. J Pediatr Surg 47:2169–2174
3. Yamataka A, Ohshiro K, Okada Y, Hosoda Y, Fujiwara T, Kohno S, Sunagawa M, Futagawa S, Sakakibara N, Miyano T (1997) Complications after cyst excision with hepaticoenterostomy for choledochal cysts and their surgical management in children versus adults. J Pediatr Surg 32:1097–1102
4. Michels N (1951) The hepatic, cystic and retroduodenal arteries and their relations to the biliary ducts. Ann Surg 133:503–524
5. Tsuchiya R, Eto T, Harada N, Yamamoto K, Matsumoto T, Tsunoda T, Yamaguchi T, Noda T, Izawa K (1984) Compression of the common hepatic duct by the right hepatic artery in intrahepatic gallstones. World J Surg 8:321–326
6. Agur AMR, Dalley AF II (2012) Abdomen-Biliary ducts. In: Agur AMR, Dalley AF II (eds) Grant's atlas of anatomy, 13th edn. Lippincott Williams & Wilkins, Philadelphia, pp 160–164
7. Baek YH, Choi SR, Lee JH, Kim MJ, Kim YH, Roh YH, Roh MH (2008) Obstructive jaundice due to compression of the common bile duct by right hepatic artery originated from gastroduodenal artery. Korean J Gastroenterol 52:394–398
8. Todani T, Watanabe Y, Toki A, Ogura K, Wang ZQ (1998) Coexisting biliary anomalies and anatomical variants in choledochal cyst. Br J Surg 85:760–763
9. Lal R, Behari A, Hari RH, Sikora SS, Yachha SK, Kapoor VK (2013) Variations in biliary ductal and hepatic vascular anatomy and their relevance to the surgical management of choledochal cysts. Pediatr Surg Int 29:777–786
10. Diao M, Li L, Cheng W (2013) Role of laparoscopy in treatment of choledochal cysts in children. Pediatr Surg Int 29:317–326
11. Nennstiel S, Weber A, Frick G, Haller B, Meining A, Schmid RM, Neu B (2015) Drainage-related complications in percutaneous transhepatic biliary drainage: an analysis over 10 years. J Clin Gastroenterol 49(9):764–770